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Trigeminal trophic syndrome secondary to recurrent meningioma

To the Editor: A 45-year-old man with a history of childhood acute lymphocytic leukemia and radiation-induced bilateral recurrent meningiomas requiring resection in 2001 and resection and Gamma-Knife in 2010 presented with a 4-week history of unexplained facial ulcerations followed by the development of headaches. He reported waking with “bloody lesions” on his face, which were preceded by facial pain he rated on a scale of 1 to 10 as an 8. He denied pruritus and picking his face. His headaches were associated with left-sided photophobia and facial numbness. Physical exam revealed a red, ulcerated, indurated left tragus. Scattered 5- to 10-mm annular pink atrophic scars with peripheral hyperpigmentation were seen on the left cheek, forehead, and nose (Fig 1). A 2- x 1-cm superficial ulceration within a well-healed surgical scar and a 1.2-cm x 6-mm crescentic superficial ulceration on the left lateral nose were noted. Dried blood was noted under his fingernails. Neurologic exam revealed diminished sensation to touch, pinprick, and temperature on the left side of the face.

Complete blood count with differential, complete metabolic profile, and liver function tests were normal. Skin biopsy of the left tragus showed normal skin without evidence of malignancy or significant inflammation. Brain magnetic resonance imaging study showed a left middle cranial fossa meningioma with interval enlargement compared with the previous year with involvement of the ipsilateral trigeminal nerve (Fig 2). A right posterior clinoid meningioma, unchanged from the previous study,



Fig 1. Trigeminal trophic syndrome. Left facial erosions, scarring, and ulceration of the tragus.



Fig 2. Axial T1-weighted magnetic resonance imaging of the brain. Left- and right-sided meningiomas (black arrowheads) and trigeminal nerves (white arrowheads).

was also present. Trigeminal trophic syndrome (TTS) involving all 3 branches of the trigeminal nerve was diagnosed.

TTS, also called trigeminal neurotrophic ulceration, is a rare condition occurring after damage to the trigeminal nerve and presents with unilateral anesthesia with crescentic ulceration classically involving the ala nasi. Ulceration is self-induced secondary to paresthesias and patients are often unaware of self-mutilation. Causes of trigeminal nerve damage leading to TTS include trigeminal ablation, infarction, vertebrobasilar insufficiency, acoustic neuroma, astrocytoma, meningioma, spinal cord degeneration, *Mycobacterium leprae* neuritis, herpes zoster, syringobulbia, postencephalitic parkinsonism, and trauma.¹

Lesions of TTS most commonly occur in the second trigeminal division dermatome, followed by the first and third.² One case of TTS involving all 3 branches of the trigeminal nerve in the context of surgical resection for meningioma has been reported.³ Another report described a patient in whom trigeminal sensory dysfunction developed 1 year after meningioma excision and subsequently ala nasi ulceration developed 6 weeks after a third surgery.⁴

Trigeminal dysfunction causing facial numbness was reported in 11% of patients who underwent surgery for cranial base meningiomas.⁵ Nerve injury was attributed in some patients to intraoperative dissection within the cavernous sinus, tumor infiltration, or gross injury to the nerve distal to the cavernous sinus; 42% of patients had no identifiable cause. We report a case of TTS involving V1-V3 related to a recurrent meningioma. Although our patient had a history of surgical resection and Gamma-Knife therapy, which could have caused trigeminal nerve damage, the association with additional neurologic symptoms and the evidence visualized on neuroradiologic imaging suggest recurrent disease as the etiology of the TTS. Interestingly, the appearance of skin lesions preceded the development of headaches, suggesting that TTS can indicate neurologic disease before the development of other neurologic symptoms.

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Striking leflunomide efficacy against refractory cutaneous sarcoidosis

To the Editor: Sarcoidosis, a multisystemic granulomatous disease of unknown etiology, involves the skin in about 25% of cases.¹ We describe leflunomide efficacy in 2 patients whose cutaneous sarcoidoses were refractory to conventional therapies.

Patient 1 was a 46-year-old woman with lung (stage 1), skin, and nasosinusoidal sarcoidosis lasting 16 years. Her medical history included hypertension and thalidomide-induced lower limb neuropathy. Skin lesions were resistant to topical steroids, tacrolimus, hydroxychloroquine, thalidomide, and methotrexate; the patient refused oral prednisone. Leflunomide (20 mg/day) was started alone because she had progressive nasosinusoidal involvement and cutaneous lesions on her limbs and buttocks, and a large patch on her right cheek (Fig 1, A). Angiotensin-converting enzyme (ACE) level was 84 (normal <41 IU/mL). Dramatic cutaneous improvement was noted at 6 months, and complete remission (CR) of cutaneous (Fig 1, B) and nasosinusoidal lesions was obtained after 1 year of leflunomide therapy as shown by normal ACE level and normal control skin biopsy of the left arm. Because sarcoid joint manifestations occurred whenever the leflunomide dose was decreased to less than 10 mg/day, maintenance therapy with that dose was pursued without any relapse after 34 months. Vulvar and cervical intraepithelial neoplasias, diagnosed after 24 months, were treated with surgical excision and cervical conization, respectively, without recurrence 1 year later.

Patient 2, a 51-year-old woman with type 1 diabetes and hypertension had stage 1 lung and nasosinusoidal sarcoidosis and severe facial lupus pernio (Fig 2, A) lasting 9 years that was resistant to topical and systemic corticosteroids, thalidomide, infliximab, etanercept, efalizumab, methotrexate alone, methotrexate combined with hydroxychloroquine, and intralesional corticosteroid injections. Leflunomide (20 mg/day) was added to a stable methotrexate dose (20 mg/wk). Striking regression of skin lesions and nasosinusoidal symptoms was obtained after 4 months of leflunomide therapy (Fig 2, B). Discontinuing methotrexate and